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A Systematic Review of the Economic Evaluation of Telemedicine in Japan

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Objectives: There is no systematic review on economic evaluations of telemedicine in Japan, despite over 1000 trials implemented. Our systematic review aims to examine whether Japan's telemedicine is cost-saving or cost-effective, examine the methodological rigorousness of the economic evaluations, and discuss future studies needed to improve telemedicine's financial sustainability.

Methods: We searched five databases, including two Japanese databases, to find peer-reviewed articles published between January 1, 2000 and December 31, 2014 in English and Japanese that performed economic evaluations of Japan's telemedicine programs. The methodological rigorousness of the economic analyses was assessed with a well-established checklist. We calculated the benefit-to-cost ratio (BCR) when a reviewed study reported related data but did not report the BCR. All cost values were adjusted to 2014 US dollars.

Results: Among the 17 articles identified, six studies reported on settings connecting physicians for specialist consultations, and eleven studies on settings connecting healthcare providers and patients at home. There are three cost-benefit analyses and three cost-minimization analyses. The remaining studies measured the benefit of telemedicine only, using medical expenditure saved or users' willingness-to-pay. There was substantial diversity in the methodological rigorousness. Studies on teleradiology and teledermatology indicated a favorable level of economic efficiency. Studies on telehomecare gave mixed results. One cost-benefit analysis on telehomecare indicated a low economic efficiency, partly due to public subsidy rules, e.g., a too short budget period.

Conclusions: Overall, telemedicine programs in Japan were indicated to have a favorable level of economic efficiency. However, the scarcity of the economic literature indicates the need for further rigorous economic evaluation studies.

Key words: Telemedicine, Telehealth, Economic evaluation, Cost-benefit analysis

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INTRODUCTION

Telemedicine, broadly defined as the use of medical information exchanged from one site to another via electronic communications to improve a patient's clinical health status [1], has been expected to improve simultaneously the cost of, quality of, and access to healthcare. There are over 80 systematic reviews on the effectiveness of telemedicine and more than 20 of them reporting conclusively that telemedicine is effective [2] in such areas as mental health [3-5] and management of chronic diseases [6-8] including diabetes, heart failure, and elderly care [9,10].

However, previous systematic reviews on the economic evaluation of telemedicine, based on study populations in the US, the UK, and Australia, show there is no conclusive evidence that telemedicine interventions are cost-saving or cost-effective compared to conventional health care [11-14]. In many countries, it has been reported that a large number of telemedicine programs have been started on a trial basis but discontinued for larger scale implementation due to lack of sustainable financial sources [15-19]. This is mainly because most insurers do not reimburse new types of healthcare delivery [20-22]. For this reason, several studies have stated that comprehensive cost-effectiveness studies are essential in justifying insurer coverage and reimbursement for telemedicine in the future [23,24].

Japan has faced a growing proportion of elderly people amidst a declining overall population. In 2014, the proportion of the population aged 65 or older was estimated to be 25.9%—the highest proportion in the world [25]. Currently, two major healthcare issues in Japan are the increasing total cost of healthcare and the uneven distribution of healthcare resources. The latter issue limits the access of remotely located people to sufficient healthcare services. Telemedicine is deemed to be a potential solution for these current twofold issues.

Based on the budget records issued by Japan's Ministry of Finance, we calculated public expenditure for development and pilot trials of health information technology, including telemedicine, from 2008 to 2014—the result was \$1.5 billion. Despite such enormous amounts in public subsidies for telemedicine, the literature indicated serious concern about the financial sustainability of telemedicine [26,27].

Among the studies reporting the effectiveness of telemedicine experiments around the country, only a small subset of these studies conducted an economic evaluation. The absence of a cohesive body of rigorous economic evaluation studies may be one of the key obstacles to the widespread adoption, proliferation, and funding of telemedicine programs [9,28-30].

This study's aim was to conduct a systematic review to identify and analyze the published economic evaluations on telemedicine in Japan. This qualitative assessment has three objectives: (a) to identify whether, and in what areas, telemedicine was evaluated to achieve a favorable level of economic efficiency, that is, whether it was cost-saving or cost-effective in Japan; (b) to assess the methodological rigor of the economic evaluations; and (c) to discuss future studies neces-

sary to improve the general financial sustainability of telemedicine.

METHODS

This article focuses on telemedicine in broader health areas to make the results more comprehensive. Specifically, this study gathered economic evaluations of telemedicine connecting “non-specialized physicians and specialized physicians,” “physicians and patients,” and “public health nurses and patients.”

Search Strategy

We searched the following databases: PubMed/MEDLINE, Web of Science, IEEE Xplore, Ichushi-Web database (the largest Japanese health database), and CiNii Articles (the largest Japanese database in all academic fields). All peer-reviewed articles published between January 1, 2000 and December 31, 2014 in English and Japanese were selected if they included any type of economic evaluation in monetary terms.

The inclusion criteria regarding economic evaluation were full economic evaluation [31] (i.e., cost-minimization analysis [CMA], cost-effectiveness analysis [CEA], cost-utility analysis [CUA], cost-benefit analysis [CBA]), cost-only analysis, or benefit-only analysis. The exclusion criteria were articles that described only effectiveness or benefit in non-monetary terms and articles available only in abstract form. Table 1 shows the different and exact combinations of keywords, relating to economic evaluation and telemedicine, used in this research. We used comparable Japanese words for Japanese databases.

Review Process

The selection process for the articles involved reading the titles and abstracts of the results obtained by one of the authors. After duplicate articles were deleted, all abstracts were read for relevance based on the inclusion and exclusion criteria. The relevant full-text articles were then obtained and thoroughly read by two authors. A classification of the telemedicine types, economic-analysis types, study designs, study populations, time frames, and the selection for the final appraisal were obtained by reading both the abstracts and the entire articles. In case of any disagreement, the two authors discussed the article and one designated author made the final decision.

In order to compare a variety of studies conducted in different time periods, we made a purchasing power parity adjust-

Table 1. Search strategy

Database ¹	Keywords for search
PubMed/MEDLINE	("costs and cost analysis"[MeSH terms] OR "cost benefit analysis"[MeSH terms] OR "cost effectiveness" OR "cost utility") AND ("telemedicine"[MeSH terms] OR telecare OR telehealth OR e-health) AND Japan
Web of Science	TS=("cost\$analysis" OR "cost*benefit" OR "cost*effectiveness" OR "cost*utility" OR "economic evaluation") AND TS=(telemedicine OR tele*care OR tele*health OR e*health) AND TS=(Japan*)
IEEE Xplore	("cost analysis" OR "cost benefit" OR "cost effectiveness" OR "cost utility" OR "economic evaluation") AND (telemedicine OR telecare OR telehealth OR e-health) AND Japan
Ichushi-Web ²	(in Japanese) Within "Original Article" category, used ("cost analysis" OR "cost benefit" OR "cost effectiveness" OR "cost utility" OR "economic evaluation") AND (telemedicine OR telecare OR telehealth OR e-health)
CiNii Articles ²	(in Japanese) ("cost analysis" OR "cost benefit" OR "cost effectiveness" OR "cost utility" OR "economic evaluation") AND (telemedicine OR telecare OR telehealth OR e-health)

¹The index period was 2000-2014 except for IEEE Xplore, which only allowed a search of journals published after 2002.

²For Japanese databases, relevant keywords were used, and the word "Japan" was not used.

ment and converted all costs to 2014 US dollars using the consumer price index [25].

Economic Evaluation Quality Assessment

We critically assessed the methodological rigorousness of the economic evaluation for each article based on the latest version of the checklist developed by Drummond et al. [31]. This checklist was chosen partly because it was one of the most widely used assessment tools in this area, e.g., this checklist's older version was known and widely used as "the BMJ Check-List" [32,33]. The most recent version of this checklist, published in 2015 [31], contains 87 items under ten headings (detailed in Table 2). Assessors are asked to check either "yes," "no," or "not applicable" (N/A) for each of ten subheadings according to the relative importance subjectively conferred to each item under each subheading. There is no explicit weighting for each item to calculate a single aggregated score. This checklist concentrates on full economic evaluations, but could also be used for partial economic evaluations, reports, and commentaries on economic evaluations. If items were not applicable to a specific study, a "N/A" response could be used.

Miki Akiyama and Byung-Kwang Yoo graded three studies using the criteria in order to reach consensus on the interpretation of the criteria, and Miki Akiyama graded the remaining papers.

Supplemental Estimation Based on Published Data

When a reviewed study reported cost and benefit but did not report the benefit-to-cost ratio (BCR) [34-36], we made a supplemental estimation of BCR by dividing the reported financial benefit by the reported cost [31]. The financial benefit

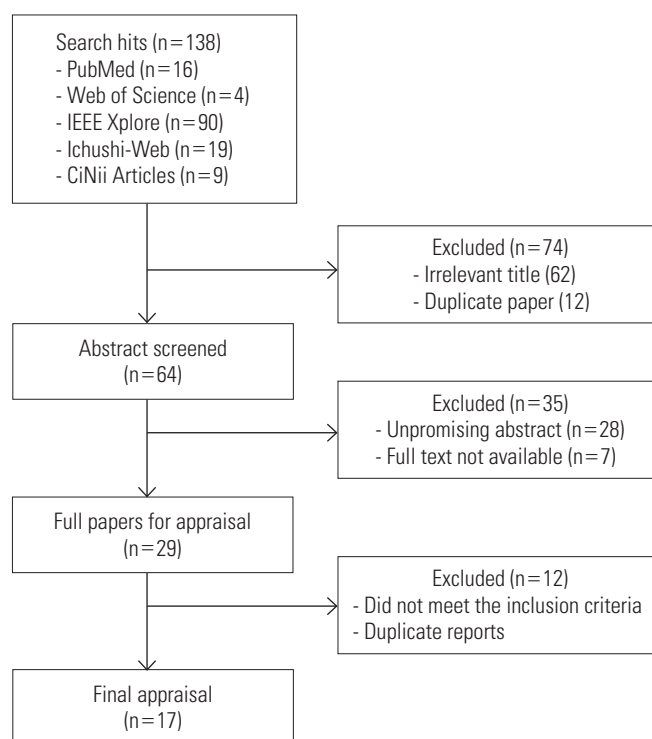


Figure 1. Flowchart of the steps followed in this review.

was estimated as the reduced medical care expenditure without assigning any monetary value for an improvement in health outcome.

RESULTS

Summary of Identified Literature

Figure 1 shows a flowchart with the steps followed in this review. The initial screening of the electronic databases retrieved 138 titles and abstracts; after the removal of duplicates

Table 2. A checklist for assessing the quality of economic evaluations¹ [31]

Contain items
1. Was a well-defined question posed in answerable form? 1.1. Did the study examine both costs and effects of the service(s) or programme(s) over an appropriate time horizon? 1.2. Did the study involve a comparison of alternatives? 1.3. Was a viewpoint for the analysis stated and was the study placed in any particular decision-making context? 1.4. Where the patient population and any relevant subgroups adequately defined?
2. Was a comprehensive description of the competing alternatives given (i.e. can you tell who did what to whom, where, and how often)? 2.1. Were there any important alternatives omitted? 2.2. Was (should) a do-nothing alternative (be) considered? 2.3. Were relevant alternatives identified for the patient subgroups?
3. Was the effectiveness of the programme or services established? 3.1. Was this done through a randomised, controlled clinical trial? If so, did the trial protocol reflect what would happen in regular practice? 3.2. Was effectiveness established through an overview of clinical studies? If so, were the search strategy and rules for inclusion or exclusion outlined? 3.3. Were observational data or assumptions used to establish effectiveness? If so, were any potential biases recognized?
4. Were all the important and relevant costs and consequences for each alternative identified? 4.1. Was the range wide enough for the research question at hand? 4.2. Did it cover all relevant viewpoints? (Possible viewpoints include the community or social viewpoint, and those of patients and third-party payers. Other viewpoints may also be relevant depending upon the particular analysis.) 4.3. Were the capital costs, as well as operating costs, included?
5. Were costs and consequences measured accurately in appropriate physical units (e.g. hours of nursing time, number of physician visits, lost work-days, gained life years)? 5.1. Were the sources of resource utilization described and justified? 5.2. Were any of the identified items omitted from measurement? If so, does this mean that they carried no weight in the subsequent analysis? 5.3. Were there any special circumstances (e.g., joint use of resources) that made measurement difficult? Were these circumstances handled appropriately?
6. Were the cost and consequences valued credibly? 6.1. Were the sources of all values clearly identified? (Possible sources include market values, patient or client preferences and views, policy-makers' views and health professionals' judgements) 6.2. Were market values employed for changes involving resources gained or depleted? 6.3. Where market values were absent (e.g. volunteer labour), or market values did not reflect actual values (such as clinic space donated at a reduced rate), were adjustments made to approximate market values? 6.4. Was the valuation of consequences appropriate for the question posed (i.e. has the appropriate type or types of analysis – cost-effectiveness, cost-benefit, cost-utility – been selected)?
7. Were costs and consequences adjusted for differential timing? 7.1. Were costs and consequences that occur in the future 'discounted' to their present values? 7.2. Was there any justification given for the discount rate used?
8. Was an incremental analysis of costs and consequences of alternatives performed? 8.1. Were the additional (incremental) costs generated by one alternative over another compared to the additional effects, benefits, or utilities generated?
9. Was allowance made for uncertainty in the estimates of costs and consequences? 9.1. If patient-level data on costs or consequences were available, were appropriate statistical analyses performed? 9.2. If a sensitivity analysis was employed, was justification provided for the form(s) of sensitivity analysis employed and the ranges or distributions of values (for key study parameters)? 9.3. Were the conclusions of the study sensitive to the uncertainty in the results, as quantified by the statistical and/or sensitivity analysis? 9.4. Was heterogeneity in the patient population recognized, for example by presenting study results for relevant subgroups?
10. Did the presentation and discussion of study results include all issues of concern to users? 10.1. Were the conclusions of the analysis based on some overall index or ratio of costs to consequences (e.g. cost-effectiveness ratio)? If so, was the index interpreted intelligently or in a mechanistic fashion? 10.2. Were the results compared with those of others who have investigated the same question? If so, were allowances made for potential differences in study methodology? 10.3. Did the study discuss the generalisability of the results to other settings and patient/client groups? 10.4. Did the study allude to, or take account of, other important factors in the choice or decision under consideration (e.g. distribution of costs and consequences, or relevant ethical issues)? 10.5. Did the study discuss issues of implementation, such as the feasibility of adopting the 'preferred' programme given existing financial or other constraints, and whether any freed resources could be redeployed to other worthwhile programmes? 10.6. Were the implications of uncertainty for decision-making, including the need for future research, explored?

¹Assessors are asked to check either "yes," "no," or "not applicable" for each of the 10 subheadings according to the relative importance subjectively conferred to each item under each subheading.

and irrelevant titles, 64 abstracts remained for screening and 29 potentially eligible full-text articles were obtained. We excluded 12 articles after reading the full text, which were judged to meet the exclusion criteria. Two were general reviews of the situation in Asia, one was a review on the methodology of economic evaluation, four were benefit analyses using the same survey data, and five papers were not peer-reviewed. Eventually, 17 peer-reviewed articles containing any form of economic appraisal in monetary terms remained.

Table 3 summarizes the characteristics and key findings of the selected studies. Among these studies, 12 were published in English [34-45] and five in Japanese [27,46-49]. The service settings of telemedicine were categorized into two major types: those connecting physicians for specialist consultations (the MD-MD setting) [34,35,37-39,49] and those connecting healthcare providers and patients [27,36,40-48], which were further sub-categorized into settings connecting physicians and patients at home (the MD-Patient setting), and those connecting public health nurses and patients at home (the PhNrs-Patient setting).

Clinical disciplines within MD-MD settings included dermatological consultation [35], pediatric medical consultation [49], emergency radiotherapy for malignant spinal cord compression [34], real-time computed tomography (CT) scan indication [37], teleradiology, and telepathology [38,39].

All of the 11 settings connecting healthcare providers and patients—that is, the MD-Patient setting and PhNrs-Patient setting—dealt with home-based systems with two types of functions. One function type is a stand-alone real-time video conference system for palliative care for cancer patients without continuous monitoring of vital signs [36]. The other type, evaluated by 10 articles [27,40-48], monitors vital signs with equipment such as an automated sphygmomanometer and a simple electrocardiograph for home. Healthcare providers contact a patient when an algorithm for monitored data detects an abnormal vital sign. This type of system targeted lifestyle-related diseases such as high blood pressure, diabetes, stroke, and heart failure among fragile elderly patients.

Regarding the types of economic evaluation, they were classified into four types; three were CBA [34,40,48], which provide the most useful policy implication, that is, benefit measured by monetary value compared to input cost of telemedicine. Three studies were categorized as CMA [35-37], which estimate the cost reduction by telemedicine compared to a control group—both telemedicine and control groups achieved

the same level of health outcome. Four studies [27,43-45] measured medical expenditure saved due to telemedicine, without calculating the input telemedicine cost or showing equal health outcomes. The remaining seven studies [38,39,41,42,46,47,49] measured willingness-to-pay (WTP) for using telemedicine. There were no CEA or CUA studies.

MD-MD Specialist Consultation

Out of six studies of MD-MD settings, one clinical efficacy study presented detailed estimates of both the costs and benefits of telemedicine in emergency radiotherapy for malignant spinal cord compression (MSCC), connecting between a university hospital and 10 rural hospitals [34]. Since this study did not report the BCR, we calculated the ratios to range from 4.67 to 13.40.

There were two CMA studies regarding MD-MD specialist live consultations. One CMA study on dermatology consultation [35] reported cost-saving of \$330 per person per week. This study's two-way sensitivity analysis indicated that the cost-saving results were sensitive to the two key parameters of travel time and consultation time. Assuming that travel time was 60 minutes and consultation time was five minutes (based on expert opinions from Japanese dermatologists), we calculated the BCR to be 1.53.

Another CMA study [37] examined teleradiology diagnosis accuracy in an MD-MD setting. This study estimated that teleconsultation contributed to cost-saving of \$144 per reading by avoiding unnecessary enhanced CT scans.

One survey [49] estimated the benefits of specialist teleconsultation in pediatric emergency care for primary care physicians in rural, underserved areas. These benefits were measured by inquiring about WTP among parents of children in two rural towns, that is, \$31.40 to \$33.60 per emergency department visit.

There were two studies conducted by one study group to estimate medical providers' WTP for teleradiology and telepathology [38,39]. These two studies applied different statistical methods for the same set of survey data obtained from healthcare providers across Japan, who used either teleradiology or telepathology in an MD-MD setting. Their WTP estimates were \$489 to \$510 for teleradiology and \$1063 to \$1111 for telepathology per person per year.

Healthcare providers-patient homecare

One CMA study [36] on home-palliative care in an MD-Pa-

Table 3. Summary of studies

Author (published year) [Ref] language	Service setting & location	Clinical discipline	Study design	Econ Eval type	Quality asses ¹	Perspec- tive	Time horizon (discount) depreciation	Sample size	Cost measurement	Consequence measurement	Key findings (SA)	Funding source
Hashimoto et al. (2001) [34] ENG	MD-MD live consultation rural hospitals	Emergency radiotherapy	ProsCoh	CBA	6/3	Health care system	1 y (no need) Not specified	1073	Initial system and operational costs for one university hospital and 10 rural hospitals including consultation fee	Recovery rate of non-ambulant patients, % of avoiding hospitalization	Cost-saving 4.67 < BCR < 13.4 SA: not performed	Not specified
Takada et al. (2003) [37] ENG	MD-MD live consultation rural hospitals	Diagnostic radiology	DiagAcc	CMA	6/3	Health provider	1 y (no need) Not specified	500	Only cost of films used for diagnosis and consultation fees are shown No initial cost shown	Diagnosis accuracy, % of avoiding unnecessary enhanced CT	Cost-saving (\$144 per reading) SA: not performed	Not specified
Dekio et al. (2010) [35] ENG	MD-MD live consultation rural hospitals	Dermatology	DiagAcc	CMA	9/1	Health provider	1 wk (no need) Not specified	107	Initial system cost and monthly costs including medical staff's fees, travel costs, and overhead office fee	Screen image diagnoses, travel time and cost saved	Cost-saving BCR = 1.53 (when travel/consultation time = 60 min/ 5 min) (\$330 saving per case per week) ² SA: 2-way	MEXT
Tanikawa et al. (2010) [49] JPN	MD-MD live consultation rural clinics	Pediatric primary care	Survey	WTP estimation	2/7	Societal	1 d (no need) Not specified	263	N/A	Patients' WTP for telemedicine use between remote primary pediatrician and university hospital	WTP = \$31-34 per emergency department visit SA: not performed	Not specified
Miyahara et al. (2006) [38] ENG	MD-MD stored nation-wide	Teleradiology, telepathology	Survey	WTP estimation	1/8	Health provider	1 y (no need) Not specified	622	N/A	Medical institutions' WTP for teleradiology, pathology, consultation (MD-MD)	WTP = \$489 per person per year for teleradiology, \$1063 for telepathology SA: not performed	MEXT, Postal Saving Funds
Tsuji et al. (2006) [39] ENG	MD-MD stored nation-wide	Teleradiology, telepathology	Survey	WTP estimation	1/8	Health provider	1 y (no need) Not specified	622	N/A	Medical institutions' WTP for teleradiology, pathology, consultation (MD-MD)	WTP = \$510 per person per year for teleradiology, \$1111 for telepathology SA: not performed	JSPS, MHLW, Okawa Telecom Foundation, Postal Saving Funds
Aoki et al. (2006) [36] ENG	MD-Pt live rural home	Palliative care	RetCoh	CMA	9/0	Health care system	1 y (no need) 5 y depreciation	100	Initial system cost and monthly costs including medical staff's fees and travel costs.	Patients' satisfaction (qualitative), travel time and cost saved	BCR = 1.93 (\$5000 saving per case per year) ³ SA: 1-way and 2-way	MHLW

(Continued to the next page)

Table 3. Continued from the previous page

Author (published year) [Ref] language	Service setting & location	Clinical discipline	Study design	Econ Eval type	Quality assess ¹	Perspec- tive	Time horizon (discount) depreciation	Sample size	Cost measurement	Consequence measurement	Key findings (SA)	Funding source
Tsuji et al. (2002) [48] JPN	MD-Pt, stored rural home	Elderly with chronic diseases	RetCoh	CBA	8/2	Societal	6-11 y (4%) 6 y depreciation	405	Initial system and device cost and operational costs including labor fees.	Users' health consciousness, anxiety, and WTP for telehomecare service	Cost-saving BCR (6 y) = 1.07 BCR (11 y) = 1.28 SA: not performed	MHLW
Tsuji et al. (2003) [40] ENG	MD-Pt, stored rural home	Elderly with chronic diseases	RetCoh	CBA	8/2	Societal	6 y (4%) 6 y depreciation	2333	Initial system and device cost and operational costs including labor fees.	Users' health consciousness, anxiety, and WTP for telehomecare service	BCR in 4 regions ranged from 0.54 to 1.07 SA: not performed	MEXT
Ito et al. (2003) [46] JPN	MD-Pt, stored rural home	Elderly with chronic diseases	Survey	WTP estimation	2/7	Societal	1 mo (no need) Not specified	498	N/A	Users' health consciousness, anxiety, and WTP for telehomecare service	WTP = \$288 per case per year for telehomecare; no difference between 2 towns after adjustment SA: not performed	Not specified
Osaka et al. (2003) [47] JPN	MD-Pt, stored rural home	Elderly with chronic diseases	Survey	WTP estimation	1/8	Societal	1 mo (no need) Not specified	348	N/A	Users' health consciousness, anxiety, and WTP for telehomecare service	WTP = \$480 per case per year for telehome care SA: not performed	Not specified
Tsuji et al. (2003) [41] ENG	MD-Pt, stored rural home	Elderly with chronic diseases	Survey	WTP estimation	1/8	Societal	1 mo (no need) Not specified	348	N/A	Users' health consciousness, anxiety, and WTP for telehomecare service	WTP = \$520 per case per year for telehome care SA: not performed	MEXT
Homma et al. (2012) [42] ENG	MD-Pt, stored urban home	Elderly with chronic diseases	Survey	WTP estimation	1/8	Societal	1 mo (no need) Not specified	102	N/A	Users' WTP for monitoring service	WTP = \$109 per case per year for chronic care SA: not performed	Omron Health Care Co.
Akematsu et al. (2009) [43] ENG	PhNrs-Pt rural home	Elderly with chronic diseases	RetCoh	Benefit estimation	3/7	Societal	10 y (not applied) Not specified	862	N/A	Medical expenditure saved by telehomecare based on receipts data of NHI	Savings of \$148 per case per year for outpatient care with lifestyle- related illness SA: not performed	MHLW, MEXT, JSPS
Akematsu et al. (2010) [27] JPN	PhNrs-Pt rural home	Elderly with chronic diseases	RetCoh	Benefit estimation	3/6	Societal	1 y (no need) Not specified	862	N/A	Medical expenditure saved by telehomecare based on receipts data of NHI	Savings \$212-371 per case per year for outpatient care with heart disease, high blood pressure, and diabetes SA: not performed	Not specified

(Continued to the next page)

Table 3. Continued from the previous page

Author (published year) [Ref] language	Service setting & location	Clinical discipline	Study design	Econ Eval type	Quality asses ¹	Perspec- tive	Time horizon (discount) depreciation	Sample size	Cost measurement	Consequence measurement	Key findings (SA)	Funding source
Akematsu et al. (2012) [44] ENG	PhNrs-Pt rural home	Elderly with chronic diseases	RetCoh	Benefit estimation	3/5	Societal	1 y (no need) Not specified	408	N/A	Medical expenditure saved by telehomecare based on receipts data of NHI	Savings of \$247-387 per case per year for outpatient care with chronic diseases SA: not performed	Not specified
Akematsu et al. (2013) [45] ENG	PhNrs-Pt rural home	Elderly with chronic diseases	RetCoh	Benefit estimation	3/7	Societal	9 y (no need) Not specified	519	N/A	Medical expenditure saved by telehomecare based on receipts data of NHI	Savings of \$629 per case per year for outpatient care with chronic diseases SA: not performed	Not specified

Ref, reference number; Econ, economic; Eval, evaluation; SA, sensitivity analysis; ENG, English; JPN, Japanese; N/A, not applicable; NHI, National Health Insurance; BCR, benefit-to-cost ratio; CT, computed tomography; MD, medical doctor; Pt, patient; PhNrs, public health nurse; ProsCoh, prospective cohort; DiagAcc, diagnostic accuracy study; RetCoh, retrospective cohort; CBA, cost-benefit analysis; CMA, cost-minimization analysis; WTP, willingness-to-pay; MEXT, Japan's Ministry of Education, Culture Sports, Science and Technology; MHLW, Japan's Ministry of Health, Labor, and Welfare; JSPS, Japan Society for the Promotion of Science.

¹Quality assessment of Econ Eval: (total number of "yes"/total number of "no") of assessment criteria in the checklist in Table 2, (yes/no) detailed in Table 4.

²Difference between "live interactive teledermatology" and "conventional clinic."

³Difference between "telepalliative care combined homecare" and "homecare with eight physician visits per month, without telemedicine" when 10 patients treated simultaneously.

Table 4. Quality assessment of economic evaluation¹

	Reference number															
	34	37	35	49	38	39	36	48	40	46	47	41	42	43	27	44
Well-defined econ question (viewpoint, comparison)	No	No	Yes	No	No	No	Yes	Yes	Yes	No	No	No	No	No	No	No
Competing alternatives	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes
Effectiveness of the program	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Costs and consequences inclusion	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes	No	No	No	No	No	No	No
Costs and consequences measurement	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes	No	No	No	No	No	No	No
Cost and consequences value credibly	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes	No	No	No	No	No	No	No
Adjusted for timing and discount	N/A	N/A	No	N/A	N/A	N/A	N/A	Yes	Yes	N/A	N/A	N/A	N/A	No	N/A	No
Incremental analysis	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes	No	No	No	No	Yes	Yes	Yes
Allowance for uncertainty in the estimates	No	No	Yes	No	No	No	Yes	No	No	No	No	No	No	No	No	No
Study results including all issues of concern	No	No	Yes	No	No	No	Yes	No	No	No	No	No	No	No	No	N/A
# of "yes"	6	6	9	2	1	1	9	8	8	2	1	1	1	3	3	3
# of "no"	3	3	1	7	8	8	0	2	2	7	8	8	8	7	6	7

N/A, not applicable.

¹Each column corresponds to each of the sub-headings shown in Table 2.

tient setting reported that telemedicine could reduce the frequency of physician visits from twice to once a week. The cost-saving of home palliative care with telemedicine were estimated to be \$5000 per person per year compared to conventional home care. A sensitivity analysis of this study showed their results to be sensitive to two parameters, the number of user patients and duration of service provided. Based on the reported benefit, we calculated the BCR to be 1.93.

One study group published two CBA studies on the same type of tele-homecare service operated in different regions with different financial schemes: (a) one region where a monthly fee was charged and (b) three other regions where no fee was charged because of public subsidies. Under the former scheme, the BCR was calculated to increase monotonically over time, that is, 1.07 (6-year operation) and 1.28 (11-year operation), using WTP estimates among patients in this region [48]. Under the latter scheme, the BCR estimates in the three regions ranged from 0.54 to 0.61 [40], indicating that tele-homecare was not cost-saving.

There were four studies reporting medical expenditure saved by telehomecare connecting public health nurses and the elderly with chronic diseases in one region [27,43-45]. Based on the claims data, these studies showed that treatment periods were shortened by telemedicine. Their estimates of reduced medical expenditure ranged from \$148 to \$629 per case per year for lifestyle diseases [27,43-45]. However, the input costs of telehomecare were not shown in these four studies.

A single research team produced three survey studies and measured patients' WTP for a telehomecare system that combines vital-signs monitoring and doctor consultation. The WTP estimates were \$288, \$480, and \$520 per case per year, depending on three analytical methods [41,46,47]. There was only one study conducted in an urban area to estimate users' WTP for home tele-health monitoring. This study, published in 2012, showed WTP as low as \$109 per patient per year [42].

Economic Evaluation Quality Assessment

Table 4 summarizes our quality assessment of reviewed studies in terms of the ten criteria of the well-established checklist from Drummond et al. [31]. There were only four studies [35,36,40,48] that met more than eight criteria, and eleven studies met only three or fewer criteria. Studies of CBA and CMA tended to meet more criteria than studies that measured only the benefits of telemedicine. Only two studies [35,36] conducted a sensitivity analysis, and hence met the

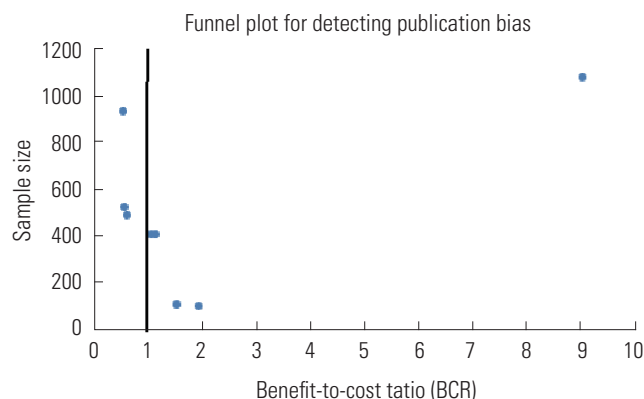


Figure 2. Funnel plot for detecting publication bias. A telehealth intervention is concluded to be cost-saving (i.e., economically efficient) when BCR (X-axis in the figure) exceeds one (represented by a bold vertical line in the figure). BCR takes on any non-negative value. A higher BCR value indicates a more economically efficient health intervention. For instance, the interpretation of BCR being two is that a \$1 investment in a telehealth intervention will generate a \$2 benefit. Due to the limited number of studies reporting BCR estimates, Figure 2 includes four plots of BCR estimates in four study sites in one publication [40]. When a reviewed paper did not report a single representative BCR estimate (e.g., mean), we calculated an average value of multiple BCR estimates.

two criteria of “uncertainty estimates” and “study results including all issues of concern.”

Publication bias

Figure 2 was created to detect a potential publication bias among the reviewed papers. A bold vertical line in this figure represents a threshold BCR value of one (also called a break-even point). A plot located rightward of this threshold vertical line indicates a favorable economic efficiency level for a telehealth program evaluated in the published study. Since the plots were skewed to the right of the threshold vertical line (i.e., BCR greater than one), Figure 2 implies a moderate bias toward publishing more studies with a higher BCR estimate for a tele-health intervention. It should be noted that our interpretation regarding such potential publication bias has limited validity due to the small number of publications with BCR estimates.

DISCUSSION

To our knowledge, this is the first systematic review to summarize the economic efficiency of Japanese telemedicine ser-

vices and to examine the methodological rigorousness of the economic evaluation studies conducted in Japan. Despite the small number and the diverse quality of economic evaluations of telemedicine, most of these evaluations indicated a favorable level of economic efficiency, that is, net cost-saving, reduced medical expenditure, and/or positive WTP.

Among studies on MD–MD live consultations in hospital settings, the most economically efficient telemedicine was emergency radiotherapy consultation about malignant spinal cord compression [34]. This study's results were consistent with another effectiveness study of spinal cord compression in Canada [50], in terms of decreasing patient transfer and shortening treatment decision time. However, the robustness of the reported BCR magnitude (>4.67) is uncertain due to the absence of sensitivity analyses.

Another cost-saving form of telemedicine in an MD–MD hospital setting was teledermatology consultation (\$17 160 per case per year) [35]. This study addressed parameter uncertainty and indicated that whether teledermatology is cost-saving or not depends on the travel time and consultation time of physicians [35]. These results do not contradict the literature reporting mixed results; that is, teledermatology cost is either lower [51] or higher [52–55] than conventional care costs.

For an MD–Patient home setting, BCR of telemedicine-combination home palliative care was estimated to be 1.93, compared to the status quo [36]. This CMA was methodologically rigorous with regard to (a) a clear statement of the purpose and perspective of the study, (b) a detailed illustration of the cost estimation, and (c) the implementation of a sensitivity analysis. This study in Japan is one of the few studies that explicitly indicated telemedicine to be cost-saving when used to supplement in-person home visits by physicians. Similar real-time, audio-visual communication services for home palliative care have been successfully used for some years in the US [56, 57] and Australia [58], while investigation in Canada [59] and pediatric telepalliative care in Australia [60] faced difficulties. In the US, the cost-saving of tele-palliative care was reported as due to a reduced number of home visits by nurses [56].

Regarding telehomecare among the elderly with chronic diseases, two CBAs reported cost-saving when implemented in one region (BCR, 1.07 to 1.28) [40,48] where patients' WTP was the only outcome measured. These studies did not account for the potential reduced medical expenditure due to telemedicine. It should be noted that an identical system deployed in three other regions with public subsidies showed

less economic efficiency (BCR ≤ 0.61) due to lower WTPs (37% to 71% of the similar one-region system above) and higher system costs (186% to 195% of the one-region system) [40]. The reason for the mixed BCR results can be partly attributed to the public subsidy rule. The restrictive rules of public subsidy are exemplified by the prohibition on carrying over the budget to a second year [40], which may have caused over-utilization of the first year cost and hence ultimately decreased BCR.

The methodological rigorousness of the reviewed studies tended to deviate from the established guidelines for economic evaluations [31,32,61–63]. For instance, only five studies [34–36,40,48] clarified the complete cost measurements of installing and operating telemedicine, which should have been compared with reduced medical expenditure due to the implementation of a telemedicine strategy such as a home telecare system (reduced by \$148 to \$629 per case per year) [27,43–45] and teleradiology (\$144 per CT reading) [37]. Therefore, other than these five studies, it seemed very difficult to determine the accurate economic efficiency level of telemedicine among the reviewed studies.

Moreover, none of the reviewed studies implemented a CEA or a CUA of the telemedicine program under focus. Therefore, it is impossible to judge whether a program is reasonably cost-effective or not when it is not cost-saving.

Another example of a common limitation among the reviewed studies is the absence or very limited application of a probabilistic analysis such as a Monte Carlo simulation in order to address the uncertainty of multiple key parameters on cost, effectiveness, and benefit, simultaneously [31].

In addition to the small number of total publications on the topic in focus, there are even fewer studies that examined comparable types of telemedicine technology and outcomes. This limited number of studies prevented us from performing a quantitative summary of the literature, such as a meta-analysis.

We believe that the following types of future studies would be useful for an international audience as well as a Japanese one. To convince policymakers to initiate and continue public subsidies for telemedicine, researchers are expected to produce more methodologically rigorous empirical evidence, presented in a reader-friendly manner. The strongest evidence is to show cost-saving based on a CBA. Due to diverse costs and benefits depending on setting, a probabilistic analysis result is useful to report; for example, cost-saving is achieved by telemedicine in 57% of 5000 iterations in emergency departments [64] and 37% of 1000 iterations in intensive care units [65].

Moreover, hospital level administrators as well as policy-makers would find it easy to understand a concrete and realistic scenario; for example, treating 10 acutely ill and injured children at a rural emergency department with telemedicine will lead to annual cost-saving of \$46 620 per emergency department [66]. These decision makers would also appreciate specific benchmarks to achieve cost-saving, such as maximum allowable cost derived from break-even analyses [65].

Since telemedicine in a probabilistic analysis is less likely to achieve cost-saving in 100% of iterations, a CEA needs to be implemented to test whether telemedicine is reasonably cost-effective or not [31]. To make CEA results of telemedicine comparable with other types of health care, a future study is expected to estimate an incremental cost-effectiveness ratio (ICER) with a unit of dollar value per additional quality-adjusted life year (QALY) by the US expert panel on cost-effectiveness in health and medicine [31,61-63]. A certain telemedicine care strategy is concluded to be cost-effective if its ICER is lower than a commonly used threshold of \$100 000 per QALY [66]. These estimates concerning CEA are also appreciated by policymakers and insurers who are interested in maximizing health outcomes under a fixed budget.

Other than improving the methodological rigorousness of economic evaluations, our review demonstrated the importance of WTP measured among users and public subsidy rules. The measured WTP could be used as a proxy for a future user charge, which is particularly important if the WTP exceeds the actual out-of-pocket payment for telemedicine. Partly because of the difference in the surveyed populations, WTP for a similar health-monitoring telemedicine care strategy declined by 75 percentage points from 2003 to 2010 [42,47]. Potential reasons for the decline include the decreased Japanese purchasing power due to long-term macro-economic recession and users' observations of continuously falling prices in other IT products and services. Thus, further studies are needed in measuring WTP under various and detailed scenarios as well as addressing the impact of public subsidy regulation on BCR. These studies could help justify increased public financial support and hence improve the financial sustainability of telemedicine programs.

CONCLUSION

In conclusion, compared to the substantial resources deployed for more than 1000 telemedicine trials in Japan, very

limited resources have been deployed for the economic evaluation of telemedicine trials. Although most reviewed economic evaluations reported a favorable level of economic efficiency, their methodological rigorousness tended to vary. Further economic evaluation studies in this field are needed with more advanced methodological rigorousness and expanded research scope (e.g., public subsidy regulation impacts). These future studies are expected to help convince various decision makers such as policymakers, insurers and hospital administrators to invest further resources in telemedicine. Consequently, the general financial sustainability of telemedicine is expected to improve in the long term.

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CONFLICT OF INTEREST

The authors have no conflicts of interest associated with the material presented in this paper.

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